CASE REPORT

# Parathyroid Double Adenoma Associated with Papillary Carcinoma of Thyroid

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## Abstract

It has been reported that thyroid disease is common in patients with hyperparathyroidism, and vice versa. Parathyroid adenoma is the most common cause of primary hyperparathyroidism (PHPT). Hypercalcemia is an important laboratory finding in the diagnosis of hyperparathyroidism. There have been sporadic reports of the coexistence of hyperparathyroidism and non-medulary carcinoma, but the association of parathyroid adenoma and papillary carcinoma of the thyroid is not well known. This is a rare report of the coexistent occurrence of papillary carcinoma of thyroid and double adenoma of parathyroid. The case is a 52-year-old woman with a 10-year history of pain in lower extremities, with initial diagnosis of osteoporosis and no clinical improvement despite using drugs. The patient was diagnosed as a case of hyperparathyroidism, using elevated concentration of calcium and PTH and metabolic bone disease (according to the isotope bone scan). In addition to parathyroid adenomas, a firm nodule in the thyroid was detected during the surgery. Therefore, lobectomy as well as hyperparathyroidectomy was performed. In pathologic examination, parathyroid double adenoma and papillary carcinoma of the thyroid were revealed.

**Keywords:** Parathyroid adenoma; Papillary carcinoma; Thyroid; Hypercalcemia

## Introduction

It has been reported that thyroid disease is common in patients with hyperparathyroidism, and vice versa.[1] Parathyroid adenoma is the most common cause of primary hyperparathyroidism. Adenomas usually involve only a single gland and the remaining glands are normal or suppressed.[2] Hypercalcemia is an important laboratory finding in the diagnosis of hyperparathyroidism.[1] Papillary carcinoma of the thyroid accounts for 50-90% of the causes of thyroid carcinoma. It occurs more often in patients aged 30-40 with approximately 3:1 the female to male ratio.[3] The most common manifestation is an asymptomatic thyroid mass or a palpable nodule in the neck. Thyroid scintigraphy (thyroid scanning) with technetium Tc99m pertechnetate (99 mTc) or radioactive iodine (iodine 131 or iodine 123) is the initial diagnostic procedure of choice for a thyroid evaluation.[4] There have been sporadic case reports of the coexistence of hyperparathyroidism and non-medulary carcinoma[1] but the association of parathyroid adenoma and papillary carcinoma of the thyroid has not been well understood.[5] The first report showing that parathyroid involvement occurs in papillary thyroid carcinoma even in the early stage of the disease was published by Tang and colleagues.[6] They found 20 cases of papillary thyroid carcinomas with parathyroid involvement among the 911 visited cases. They stated that parathyroid involvement occurs in 3 patterns: (i) direct invasion from the main tumor with an infiltrative growth, (ii) invasion of parathyroid gland with an expansive growth and formation of a pseudo-capsule between the carcinoma and the parathyroid gland, and (iii) metastatic deposition within the parathyroid gland.[6]

We have recently encountered a rare case of papillary carcinoma of the thyroid associated with double adenoma of parathyroid which was initially diagnosed to be osteoporosis. High concentration of calcium and low absorption of radio-nucleotide in the whole skeleton and ostealgia and nephrolithiasis history as well as

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elevated concentration of PTH were found. Therefore, the subject was diagnosed as a case of hyperparathyroidism. Following the report of double adenoma of parathyroid in TC-MIBI scan, the patient underwent double–adenomectomy. Thyroid carcinoma was detected coincidentally during the surgery and pathologic examination revealed a papillary carcinoma.

## Case Report

A 52-year old woman was referred to our endocrinology specialized clinic with a 10-year history of pain in the bilateral lower extremities April 2004. She also complained of early fatigue and anorexia. She denied past history of polyuria, polydypsia, nausea, vomiting, constipation, peptic ulcer, arthralgia and proximal muscle weakness. There was a 2.5–year history of using of Ca-D tablet and calcitonin spray after the initial diagnosis of osteoporosis. Despite using drugs, she did not improve clinically. In medical her history, lithotripsy performed 2 years ago and cataract surgery were mentioned. The patient said that she had metabolic disease of bone, which had been reported in a bone isotope scan performed in 21.06. 2003. It reported high absorption of radionucleotide in the whole skeleton with the most severity in calvarium and low absorption in kidneys. She disclosed urine bladder carcinoma in her mother and lung carcinoma in her brother, but thyroid malignancy was not mentioned. There was no history of neck irradiation. On physical examination, no nodule or superficial lymph node was palpable, and there was no remarkable finding except for kyphoscoliosis. Laboratory tests were reported as:

Ca: 12.5 mg/dl (8.5 – 10.5), P: 2.5 mg/dl (2.5-5.5), ALP: 140 IU/L (64-306), Hb: 10.5, ESR: 30, TSH: 0.4 mu/l. With the probability of vitamin D toxicity, all the drugs were discontinued. 2 months later; calcium concentration was 12 mg/dl and PTH was 215 pg/ml (13-54). In $^{99m}$ TC-MIBI scan, the existence of two absorption foci, one adhered to the inferior border of the thyroid's left lobe and the second one at the back of the inferior pole of the right lobe led to the diagnosis of multiple adenoma of parathyroid (Figure 1). In 27.05.2004, the patient was operated. In addition to parathyroid adenoma, a nodule (1.5 × 1 × 1 Cm) in the right lobe, a firm mass in the isthmus (1×1 Cm) which seemed to be tumorous, and two small lymph nodes were observed during the surgery. So, inferior double adenectomy, the right thyroid lobectomy and ischmectomy, and cervical lymph node dissection were performed. Post- operation concentrations of Ca and P were 8.8 and 4.4 mg/dl, respectively. In 99 Tc TCO4 scan, the left lobe of the thyroid was normal at the topmost area of the left lobe. In addition, low radionucleotide absorption was observed because of the slight remainder of the thyroid tissue. Pathologic examination revealed papillary well–differentiated carcinoma, nodular goiter with chronic thyroiditis, parathyroid double adenoma and reactive hyperplasia of lymph node without metastasis. After 3 months on 02.09.2004, she underwent a total thyroidectomy. Postoperative concentration of TSH was 0.4 mu/l with pathologic report of nodular goiter without papillary carcinoma of the thyroid.

## Discussion

Papillary carcinoma of the thyroid is the most common carcinoma of the thyroid.[3] Genetic association of hyperparathyroidism with medulary carcinoma of the thyroid is well-known, but its association with papillary carcinoma has not been well defined. For the first time, in 1956 Ogburn and Black reported one case of mixed papillary and follicular carcinoma of the thyroid associated with parathyroid adenoma in a series

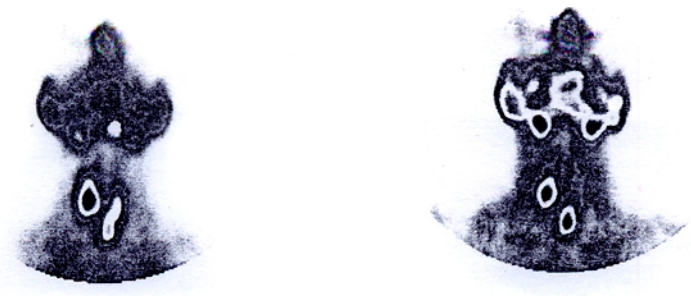

**Fig 1:** $^{99m}$ Tc MIBI isotop scan of parathyroid in a 52 years old woman shows two absorption foci, one adhered to the inferior border of the left lobe of the thyroid, and the other one at the back of the inferior pole of the right lobe, pertaining to multiple adenoma of parathyroid.

of 230 cases of hyperparathyroidism. Three out of four patients were operated primarily for parathyroid adenoma. The relationship was thought to be no more than coincidental.[7] Ellenberg *et al.* in 1962 reported five papillary, one mixed papillary and follicular in patients with hyperparathyroidism.[5]

In a study by Petro *et al.*, thyroid carcinoma was detected in five of 56 (8.9%) patients operated due to hyperparathyroidism. In only one case, a mass was palpated preoperatively. An additional 26% of the 56 patients had either multinodular goiter or follicular adenoma. It is thus important to conduct a biopsy on thyroid nodules which may represent malignancy.[8]

Linos and colleagues between 1965 and 1979 in a study performed on 2027 patients whose hyperparathyroidism was proven surgically reported 48 Grade 1 papillary carcinomas of the thyroid, 41 single adenomas of parathyroid and 5 double adenomas.[9] Sidhu and Campbell in a retrospective review of all cases of neck exploration for primary hyperparathyroidism between 1993 and 1998 found 4 cases of papillary carcinoma.[10]

Hyperparathyroidism can begin with only slight changes in bone density, function of other organs, or PTH or blood calcium levels.[1] This article is reporting a rare co-occurrence of papillary carcinoma and double adenoma of the parathyroid. By considering elevated concentration of calcium and PTH and metabolic bone disease (according to the isotope bone scan), diagnosis of hyperparathyroidism was made. In 99Tc-MIBI scan, also, double adenoma of parathyroid was reported which can account for ostalgia, nephrolithiasis, early fatigue and anorexia.

This is a case history of 52 year old woman with hyperparathyroidism in which papillary thyroid carcinoma was incidentally discovered. Coexistence of hyperparathyroidism with single adenoma of parathyroid and thyroid papillary carcinoma has been reported in some studies[6,8-10] but it has been the occurrence of double adenoma of parathyroid and coexistence of papillary thyroid carcinoma, which were incidentally discovered. Solitary parathyroid adenoma is responsible for more than 85% cases with PHPT, while 10-15% of patients demonstrate multiglandular disease. Parathyroid carcinoma is seen in less than 1% of the cases. Primary hyperparathyroidism is usually caused by a single adenoma while incidence of double adenoma ranges between 2% to 4%.[11]

In conclusion, this case report describes a patient with double adenomas of parathyroid with incidental thyroid papillary carcinoma. It is recommended that during the surgery of parathyroid adenoma, thyroid be palpated because of the probability of coincidental thyroid carcinoma.

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**Conflict of interest:** None declared.

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